Case reports

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Intractable paroxysmal tachycardia caused by a concealed retrogradely conducting Kent bundle

Demonstration by epicardial mapping and cure of tachycardias by surgical interruption of the His bundle [1]

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Electrophysiological and epicardial mapping studies are described in a patient without pre-excitation who had intractable recurrent paroxysmal supraventricular tachycardia. Electrophysiological studies revealed fixed VA conduction times during both rapid ventricular pacing and coupled ventricular stimulation. Catheter mapping of atrial activation during retrograde conduction and during induced paroxysmal supraventricular tachycardia revealed early distal coronary sinus activation (posterior left atrium) relative to the low septal, low lateral, and high lateral right atrium. These studies suggested the presence of a concealed left-sided bypass tract.

The patient underwent surgical interruption of the His bundle for control of paroxysmal supraventricular tachycardia. Epicardial mapping of the atria (during ventricular pacing) confirmed the presence of a concealed left-sided bypass tract. Surgery produced antegrade AV block (while retrograde conduction was maintained) and total cure of paroxysmal supraventricular tachycardia. This is the first reported case of a concealed retrograde extranodal pathway documented by epicardial mapping.

Recent studies in patients with re-entrant paroxysmal supraventricular tachycardia without apparent pre-excitation suggest that re-entry can either be within the AV node or involve the normal pathway and a concealed extranodal pathway (Denes *et al.*, 1973; Coumel and Attuel, 1974; Neuss, Schlepper, and Thormann, 1975; Spurrell, Krikler, and Sowton, 1974a, b; Zipes, DeJoseph, and Rothbaum, 1974). In the latter cases, the extranodal pathway is capable of only retrograde conduction and, therefore, concealed on the surface electrocardiogram. Differentiation between intra- and extranodal re-entry has both diagnostic and therapeutic implications.

In the present report, we describe the electrophysiological and epicardial mapping studies in a

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patient without ventricular pre-excitation who had recurrent, intractable supraventricular tachycardia, and who underwent surgical ablation of the His bundle. Preoperative electrophysiological studies suggested the presence of a left-sided concealed extranodal pathway capable of only retrograde conduction. Epicardial mapping of the atria at the time of cardiac surgery confirmed the presence of an anomalous left-sided connexion. After surgical section of the His bundle, complete antegrade AV block was produced with maintenance of intact retrograde conduction.

Case report

The patient was a 44-year-old male Jehovah's witness, with documented recurrent paroxysmal supraventricular tachycardia for 15 years. Electrocardiograms between attacks were within normal limits, with a PR interval
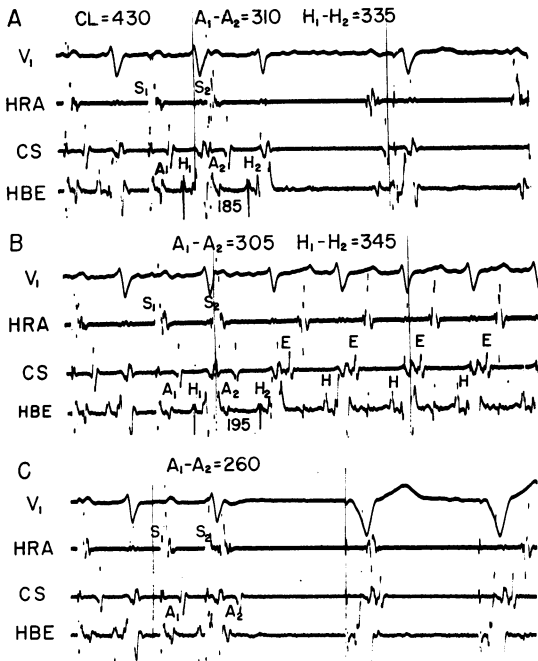
FIG. 1 *Induction of paroxysmal supraventricular tachycardia during coupled right atrial pacing at a basic cycle length of 430 ms. In panel A, the A_1-A_2 coupling interval is 310 ms. A_2 is conducted to the ventricles with an AH interval of 185 ms. In panel B, the A_1-A_2 interval has been decreased by 5 ms. A_2-H_2 interval is 195 ms and A_2 is followed by an echo beat (E) and an episode of paroxysmal supraventricular tachycardia is induced. Note that the coronary sinus (CS) electrogram of E precedes both high right atrial and low septal right atrial electrograms. In panel C, A_1-A_2 is 260 ms. A_2 is blocked in the AV node (proximal to H). The permanent ventricular pacemaker escapes following the blocked A_2.*

Because of failure of drug therapy, an attempt was made to control the tachycardia with atrial pacing, but was unsuccessful. A permanent externally activated demand epicardial ventricular pacemaker was implanted, when studies suggested that this mode of therapy would result in interruption of tachycardias. However, within several weeks of pacemaker implantation, intractable recurrent paroxysmal supraventricular tachycardia again occurred. The patient was then referred to the University of Illinois Hospital for further evaluation.

Electrophysiological studies

Electrophysiological studies were performed in January 1975, after informed consent was obtained. Medicines were discontinued 72 hours before study. His bundle electrograms were recorded with catheter techniques (Scherlag *et al.*, 1969). In addition, a quadripolar catheter was positioned in the high right atrium for recording and stimulation. A hexapolar catheter was positioned so that the distal two poles were located at the right ventricular apex, while the proximal electrodes were contiguous to the low lateral right atrium. A bipolar catheter was placed in the distal coronary sinus.

Basic conduction intervals were recorded during sinus rhythm at a cycle length of 800 ms and were as follows: PA of 42 ms, AH of 84 ms, and HV of 45 ms. With high right atrial pacing, 1:1 AV conduction was noted up to a paced rate of 200/min, with an increase in AH interval from 110 to 200 ms. AV nodal Wenckebach periods were noted at a paced rate of 210/min. There was no evidence of an antegradely conducting anomalous pathway with pacing of the low lateral right atrium, the coronary sinus, and low septal right atrium (Denes *et al.*, 1974). With right ventricular pacing at rates of 90 to 170/min VA conduction (ventricle to low septal right atrium) remained constant at 230 ms. The sequence of atrial depolarization during retrograde conduction was distal coronary sinus, preceding both low septal right atrium and high right atrium by 40 ms.

Antegrade refractory periods were measured with atrial extra-stimulus technique from the high right atrium at cycle lengths of 600 and 430 ms and are listed in the Table. The AV conduction curves (A_1-A_2, H_1-H_2) were smooth.

of 0·18 s, and a QRS duration of 0·08 s. Episodes of paroxysmal supraventricular tachycardia were characterized by rates of 170 to 190/minute and narrow QRS complexes identical to those noted during sinus rhythm.

During the two years before admission, attacks of paroxysmal supraventricular tachycardia had become more frequent and prolonged, occurring several times daily and often lasting for hours. The patient had to give up gainful employment as a lecturer because of the severity and frequency of the attacks. Therapy had included the following in high dosage, both singly and in combination: digitalis, propranolol, quinidine, phenytoin, and procainamide. The patient manifested refractoriness and/or major side effects to all of these drugs.

TABLE *Refractory periods (ms)*

CL	600	430
Atrial ERP	220	230
Atrial FRP	270	250
AV node ERP	270	260
AV node FRP	380	330
Ventricular ERP	—	280
Bypass ERP		<280
AV echo zone		305–270

CL = Cycle Length; FRP = Functional Refractory Period;
ERP = Effective Refractory Period.

Retrograde refractory period determinations were performed using the ventricular extrastimulus technique (V_2) from the right ventricular apex at a cycle length of 600 ms. V_2-A_2 remained constant at 230 ms, despite shortening of V_1-V_2 from 580 to 285 ms. The sequence of atrial activation was identical to that described above during incremental ventricular pacing. VA conduction was limited by ventricular refractoriness at an S_1-S_2 interval of 280 ms.

Both single atrial echoes and sustained paroxysmal supraventricular tachycardia were noted during pacing-induced AV nodal Wenckebach periods, with achievement of a critical AH of 260 ms (Goldreyer and Damato, 1971). An echo zone (single echoes ±episodes of sustained paroxysmal supraventricular tachycardia) was defined with atrial extrastimulus technique between A_1-A_2 coupling interval of 305 to 270 ms at a cycle length of 430 ms (Fig. 1). The critical AH for induction of echoes during coupled stimulation was 195 ms (Fig. 1B) (Goldreyer and Damato, 1971).

Induced sustained paroxysmal supraventricular tachycardia was characterized by a narrow QRS, an HV of 45 ms, and a cycle length of 367 ms. Retrograde activation times during induced paroxysmal supraventricular tachycardia were as follows (measured from the onset of the QRS to the onset of the respective atrial electrogram): V-distal coronary sinus (110 ms), V-high right atrium (145 ms), and V-low septal right atrium (160 ms) (Fig. 1B).

Short periods of functional left and right bundle-branch block were also observed during episodes of induced paroxysmal supraventricular tachycardia. The cycle length during tachycardia with left bundle-branch block was always 25 ms longer than the cycle length during narrow QRS paroxysmal supraventricular tachycardia and with functional right bundle-branch block. Ventricular extrastimuli (V_8) were coupled to the QRS (V) of the tachycardia. At V-V_8 intervals greater than 345 ms, no reset of the atrial electrogram was noted. At V-V_8 intervals between 345 and 290 ms, the corresponding A to A_8 interval progressively shortened as the V-V_8 was decreased (Fig. 2). At V-V_8 intervals between 345-325 ms, the ventricular extrastimulus (V_8) depolarized the atria (A_8) without depolarizing the His bundle (Fig. 2A). At V-V_8 between 285 to 280 ms, V_8 depolarized the atrium and interrupted the tachycardia (Fig. 2C).

The presence of a left-sided concealed extranodal pathway with ability for only retrograde conduction was suggested by the following observations: 1) absence of antegrade pre-excitation on all electrocardiograms and during atrial pacing at multiple sites (Denes *et al.*, 1974). 2) Fixed V-A intervals during incremental and coupled ventricular pacing (Wellens and Durrer, 1974a; Narula, 1974). 3) Abnormal retrograde activation sequence (with distal coronary sinus preceding all other atrial electrograms) during ventricular pacing and during paroxysmal supraventricular tachycardia (Svenson *et al.*, 1974). 4) Lengthening of cycle length of paroxysmal supraventricular tachycardia during functional left bundle-branch block (Coumel and Attuel, 1974; Spurrell *et al.*, 1974a; Neuss *et al.*, 1975). 5) Ability to capture the atria

from the ventricles during paroxysmal supraventricular tachycardia without depolarizing the His bundle (Zipes *et al.*, 1974; Neuss *et al.*, 1975). 6) Smooth antegrade AV nodal curves (Wu and Denes, 1975). During paroxysmal supraventricular tachycardia, the left-sided anomalous pathway was used for retrograde conduction and the AV node for antegrade conduction.
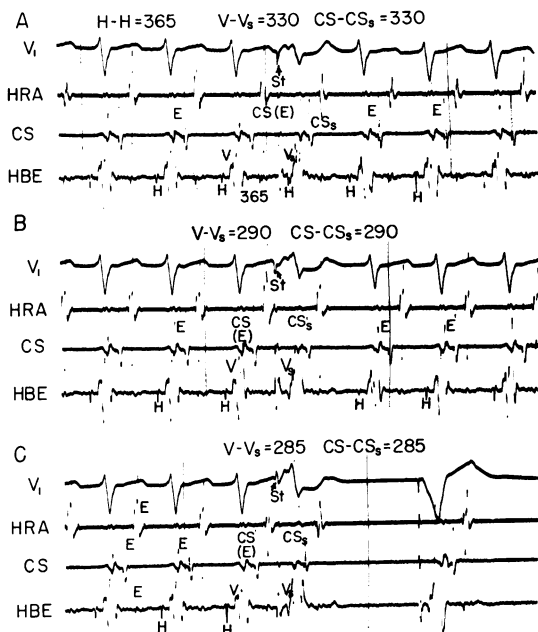

FIG. 2 *Atrial reset with premature ventricular stimulation during supraventricular tachycardia. V represents the last QRS before the introduction of the premature ventricular stimulus (V_8), CS represents the last coronary sinus electrogram before the ventricular extrastimulus. CS_8 represents the premature coronary sinus electrogram corresponding to the ventricular extrastimulus (V_8). E indicates echo and H represents the His electrogram. The cycle length of the tachycardia is 365 ms. Paper speed is 100 mm/s, time lines at 1/s.*

Panel A: The V-V_8 is 330 ms. The corresponding CS-CS_8 interval is at 330 ms. Note that the HH intervals remain undisturbed at 365 ms. This shows that an extranodal pathway must be present. Panel B: The ventricular premature beat is introduced at a V-V_8 interval of 290 ms. The corresponding CS-CS_8 interval shortens to 290 ms.

Panel C: The V-V_8 interval is decreased to 285 ms; the corresponding CS-CS_8 interval is shortened to 285 ms and blocked in the AV node. The tachycardia is then interrupted. The ventricular pacemaker escapes with retrograde conduction to the atria.

Epicardial mapping and surgery

Because of medical intractability, and because of the demonstration of a circus movement amenable to correction, the patient was referred to the Texas Heart Institute for elective transection of the His bundle. Operation was performed in March 1975. With the heart exposed, epicardial mapping of the atria was performed during right ventricular pacing using a previously described mapping technique (Svenson *et al.*, 1974; Wyndham *et al.*, 1974). The area of earliest atrial activation was noted at the lateral left atrium just adjacent to the left AV ring (Fig. 3). The patient was placed on cardiopulmonary bypass, and the His bundle was interrupted via a right atriotomy incision. Atrial pacing after closure of the right atriotomy revealed complete AV block. Ventricular pacing after closure of the atriotomy revealed intact retrograde activation. Repeat epicardial atrial mapping with ventricular pacing was identical to that before operation.

Subsequent clinical course

After operation, the patient recovered uneventfully. He has remained in heart block with pacemaker rhythm and he has been totally free of paroxysmal supraventricular tachycardia. He feels well and has resumed lecturing.

Discussion

AV re-entry can occur within the AV node or using the normal pathway and an extranodal pathway (Coumel and Attuel, 1974; Denes *et al.*, 1973; Neuss *et al.*, 1975; Spurrell *et al.*, 1974a, b; Zipes *et al.*, 1974; Wellens and Durrer, 1974a; Svenson *et al.*, 1974; Wu and Denes, 1975). The extranodal pathway can be manifest with ability for antegrade conduction (Wolff-Parkinson-White syndrome) or concealed on the surface electrocardiogram. Concealment of an extranodal pathway reflects unidirectional block with ability for only retrograde conduction. The diagnosis of a concealed extranodal pathway can be suspected by demonstration of the following electrophysiological findings: 1) absence of antegrade pre-excitation on all electrocardiograms and during atrial pacing at multiple sites (Denes *et al.*, 1974). 2) Fixed ventriculoatrial conduction times during incremental and coupled ventricular pacing (Wellens *et al.*, 1974; Narula, 1974). 3) Abnormal sequence of retrograde atrial activation (Svenson *et al.*, 1974). 4) Increase in cycle length of induced paroxysmal supraventricular tachycardia during development of functional bundle-branch block ipsilateral to the suspected anomalous pathway (Coumel and Attuel, 1974; Spurrell *et al.*, 1974a; Neuss *et al.*, 1975). 5) Retrograde capture of the atria during paroxysmal supraventricular tachycardia without discharging the His bundle (Zipes *et al.*, 1974; Neuss *et al.*, 1975). 6) Smooth antegrade conduction curves (A_1-A_2, H_1-H_2) with echo zones (Wu and Denes, 1975).

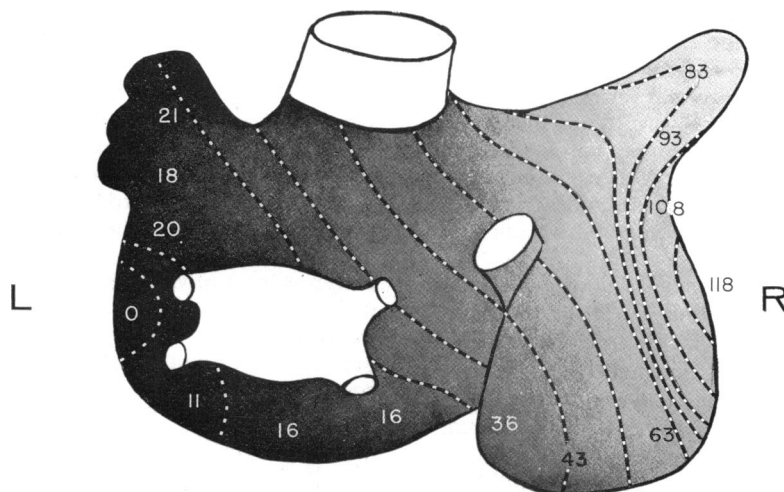All of the above electrophysiological features were shown in the present case, suggesting the presence

FIG. 3 *Posterior schematic view of epicardial atrial mapping during right ventricular stimulation.*

Isochronous lines are drawn at 10-ms intervals with reference to the earliest epicardial atrial activation (0 ms). Note that the posterolateral left atrium contiguous to the left AV ring is activated earliest.

of a concealed extranodal pathway. In addition, the findings suggested that the extranodal pathway was left-sided. The importance of our case resides in the fact that epicardial mapping of the atria confirmed the presence of a left-sided pathway, as predicted by preoperative electrophysiological studies. Epicardial mapping showed early retrograde activation of the lateral left atrium contiguous to the left AV ring.

Little is known regarding treatment of recurrent supraventricular tachycardia in patients with concealed extranodal pathways. In our experience, these cases are characterized by smooth conduction curves, as opposed to the discontinuous conduction curves often noted in patients with intra AV nodal (dual AV nodal pathway) re-entrant parosyxmal supraventricular tachycardia (Wu and Denes, 1975). Previous studies concerning induction of re-entrant tachycardia in the catheterization laboratory before and after drug administration suggest that patients with smooth conduction curves are more likely to have potentiation of induction after drugs which depress AV nodal conduction (ouabain and propranolol) (Wu et al., 1974; Wu et al., 1975). This probably reflects the fact that some of these smooth curve patients have concealed extranodal pathways underlying their paroxysmal supraventricular tachycardia (Wu and Denes, 1975). The administration of a drug depressing the AV node allows a given atrial extrastimulus to provoke greater AV nodal delays after drug administration. This additional delay potentiates the echo phenomenon, since the extranodal pathway is given more time for recovery from antegrade concealed penetration. Drugs such as procainamide and quinidine, which depress antegrade anomalous pathway conduction, should theoretically prolong retrograde refractoriness for the concealed pathway, so that these become incapable of sustaining re-entrant paroxysmal supraventricular tachycardia (Wellens and Durrer, 1974b). However, the present patient was unresponsive to procainamide and quinidine administration.

Both atrial and ventricular pacing have been used with varying degrees of success in the management of patients with intractable paroxysmal supraventricular tachycardia (Kitchen and Goldreyer, 1972). These were unsuccessful in the present case. The failure of ventricular pacing appeared to reflect the very narrow zone of coupling intervals (5 ms), in which a ventricular extrastimulus could interrupt sustained paroxysmal supraventricular tachycardia. Long periods of random ventricular pacing would be necessary before a random stimulus fell within this critical zone. Programming of a pacemaker to provide a stimulus in this zone could

also fail, since it is likely that the zone might well change in timing from day to day.

Before the use of current diagnostic electrophysiological techniques, surgical cure of intractable paroxysmal supraventricular tachycardia had been achieved by destruction of the His bundle (Gianelly et al., 1967). Surgery has also been used successfully in patients with manifest pre-excitation to interrupt re-entrant circus movements (Sealy and Wallace, 1974). Surgical ablation of anomalous pathway after demonstration with epicardial mapping can prevent recurrent circus movement paroxysmal supraventricular tachycardia (and also prevent rapid ventricular rates during paroxysmal atrial fibrillation). Similar cure of circus movement tachycardia has also been achieved in patients with pre-excitation occurring after surgical division of the normal pathway (Dreifus et al., 1968; Wellens et al., 1974).

This is the first reported case of a patient with intractable re-entrant paroxysmal supraventricular tachycardia caused by a proven concealed extranodal pathway cured by surgery. In this case, we chose to interrupt the His bundle, since this was the easiest of the possible procedures for achieving cure of the tachycardia. The fact that the patient was a Jehovah's witness and could not receive blood transfusions was another factor in reaching this decision. This operation had the disadvantage of producing chronic surgical heart block.

The other surgical approach to a case such as the present one would be to ablate surgically the concealed retrograde pathway. Theoretically, this would be ideal, since it would leave the normal conduction system intact without producing permanent heart block. However, technical difficulties might be anticipated in regard to localization of an anomalous pathway when this pathway was only capable of retrograde conduction. We believe that the feasibility of surgery for retrograde concealed pathways needs further exploration.

One additional point of interest relates to the current electrophysiological status of the present patient. He has chronic complete heart block with intact retrograde conduction. He thus resembles previously reported patients with chronic acquired complete heart block and intact retrograde conduction (Castillo and Samet, 1967; Goldreyer and Bigger, 1970). Previous authors have commented on the possibility that these patients have single lesions producing unidirectional heart block. The present case suggests that some patients with heart block and intact retrograde conduction have acquired heart block involving the normal pathway, with a concealed retrograde pathway allowing retrograde conduction.

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